Penile Paraffinoma: Should We Perform Ultrasound?☆

Parafinoma de pene: ¿le hacemos una ecografía?

Dear Editor:

The term paraffinoma refers to histological abnormality caused by subcutaneous injection of paraffin or mineral oil. The lesion is characterized by a pseudocystic pattern described in the scientific literature as *Swiss-cheese* appearance.[1] Given the intrinsic properties of this material, which is inert and cannot be absorbed by the body, it has traditionally been used to increase the volume of certain areas of the body, such as the cheeks, lips, breasts, and external genitals.[2,3]

A 42-year-old man from Romania presented with a mass, which had been growing progressively over the last 20 years, on the dorsum of the penis. The lesion had been stable until 15 days earlier, when it started to become inflamed until a painful central ulcer developed. The physical examination revealed a mass with a stony consistency measuring 5×7 cm in diameter in the dorsolateral region of the penis shaft. An ulcer of 1 cm in diameter was present on the mass, with a fibrinous base and mild serous exudate (Fig. 1a). A rounded, hard, subcutaneous tumorous mass was observed on the ventrolateral aspect of the penis, measuring 2 cm in diameter (Fig. 1b). This lesion was not painful to palpation. Of note was the presence of multiple, bilateral, swollen, stony, inguinal lymph nodes that were not painful. Given that the clinical manifestations were consistent with long-term complications of injectable filler materials, a detailed medical history was taken. This finally revealed that melted lipstick had been injected several times to increase the size of his penis.

A histopathological study revealed diffuse eosinophilic sclerosis of the reticular dermis and subcutaneous tissue, completely replacing lobules of adipose tissue while preserving the septa. Isolated foci of lymphocytic infiltrate and multiple empty cavities surrounded by giant multinucleated cells formed the typical *Swiss-cheese* image (Fig. 2).

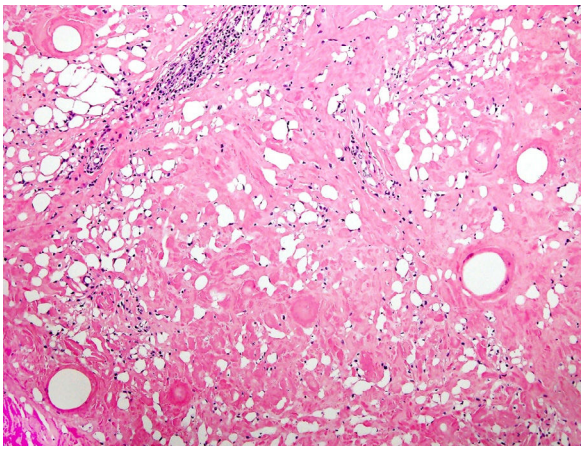

Figure 2 *Swiss-cheese* image. Diffuse sclerosis of the reticular dermis and subcutaneous tissue with empty pseudocysts.

An ultrasound study of both lesions was undertaken with a linear 14 MHz probe. The first lesion showed marked focal thickening of the subcutaneous cell tissue of the shaft of the penis, with no collections or foreign bodies detectable in the ultrasound throughout its thickness (Fig. 3a). No abnormalities were observed in the corpora cavernosa or the corpus spongiosum. In the second lesion, a well-defined hyperechogenic nodule measuring 10×12 mm in diameter was observed with a posterior acoustic shadow (Fig. 3b). In addition, bilateral hyperechogenic swollen inguinal lymph nodes with no defined fatty hila could be observed, probably related to lymphatic drainage of foreign material.

The patient underwent surgery in the urology department. The affected region was completely excised and reconstruction surgery performed with direct closure. Unfortunately, the patient refused return for lymph node

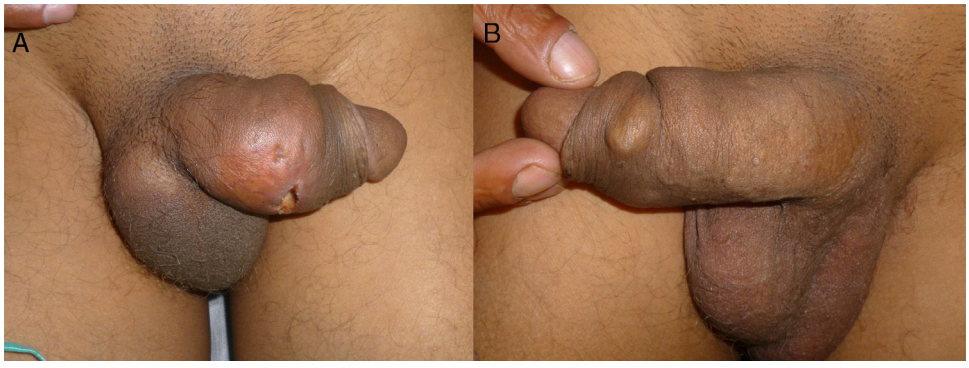

Figure 1 A, Mass of stony consistency, with ulcer with fibrinous base, on the right lateral aspect of the penis. B, Hard subcutaneous nodule, mobile, and with smooth edges, not adhered to the deep planes.

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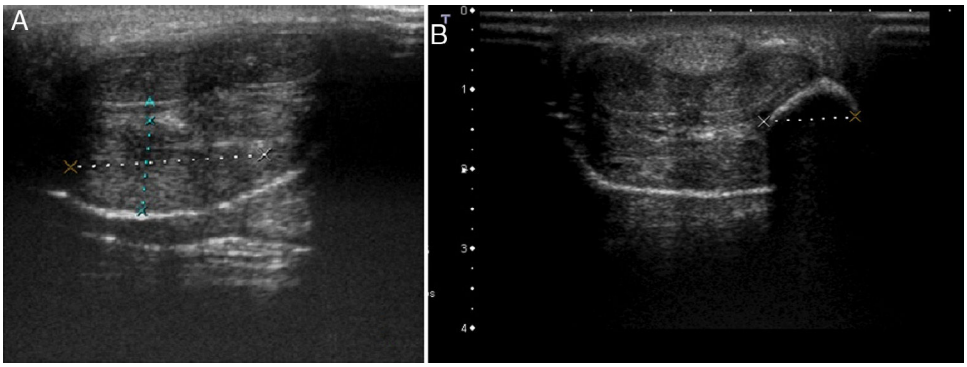

Figure 3 High-resolution ultrasound with linear 14 MHz probe. A, Marked focal thickening of subcutaneous tissue of the right ventrolateral aspect of the penis, measuring approximately 5 × 2.5 cm. Preserved corpora cavernosa. B, Oval pseudocystic lesion with marked posterior acoustic shadow.

biopsy and was definitively lost to follow-up after the operation.

Injection of paraffin or mineral oil by nonmedical personnel, with the aim of increasing the size of the penis, is a common problem in countries in Eastern Europe and certain areas of Asia.[4] Given that humans naturally lack specific enzymes to degrade mineral oils, subcutaneous injection of this type of substance is followed by a granulomatous reaction, the onset of which can be delayed by years. Clinically, onset is usually very nonspecific, with progressive induration, fluctuating edema, and/or local inflammation of the affected area which, as time passes, may be accompanied by necrosis, ulceration, and deformity.

Histology is, without doubt, key for achieving definitive diagnosis of these lesions. Typically, paraffinoma is characterized by the replacement of subcutaneous cell tissue with empty cystic spaces of variable size that give a *Swiss-cheese* appearance.[1] The process is accompanied by a granulomatous foreign body reaction, with extensive fibrosis and marked inflammatory infiltrate, mainly composed of giant multinucleated cells.[4]

Although skin abnormalities at the injection site are the most frequent complications, other potentially more serious reactions may occur, as was the case in our patient, with likely regional lymph node involvement.[5] There have even been reports of fatal lung involvement caused by migration of the filler material used.[6]

An exhaustive medical history is essential for diagnosis, as the patient may omit part of the prior history due to fear or shame.[3] Differential diagnosis should include infectious processes such as abscesses, canker sores, thrombosis of the dorsal vein of the penis, and tumors.[7] Ultrasound, which is a widely available and inexpensive technique, can be very useful for differential diagnosis. This imaging technique can also determine the involvement of structures such as the corpora cavernosa and regional lymph nodes, as well as detect the formation of abscesses or fistulas. Furthermore, it can help identify the type of filler material, as specific echographic patterns have been described for some of these.[8–10] For example, in the second image, the so called snow storm pattern is observed, where a hyperechogenic region has a posterior acoustic shadow. This pattern is typical in nonabsorbable filler materials such as silicone oil, pure silicone,[10] and, in our case, liquid paraffin.

To date, the only truly effective and definitive treatment for penile paraffinoma is surgical excision. This process may, at times, require substantial surgical reconstruction through grafts and/or plasties to cover the resulting defect.[5]

It is very important that the dermatologist recognizes the complications of filler materials, particularly when applied by nonmedical personnel. Ultrasound is of great help, both for identifying the filler material and for recognition of local complications or involvement of regional lymph nodes.

Conflicts Of Interest

The authors declare that they have no conflicts of interest.

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C. Morales-Raya,* A. Calleja-Algarra, F. Tous-Romero, R. Rivera-Díaz

Servicio de Dermatología, Hospital Universitario 12 de Octubre, Madrid, Spain

*Corresponding author. Mr. Carlos Morales-Raya Hospital Universitario Doce de Octubre Dermatología Av. de Córdoba s/n 28041 Madrid Spain. Phone: +34690325000.
E-mail address: carlosmoralesraya@hotmail.com (C. Morales-Raya).

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Facial Cutaneous Rosai-Dorfman Disease☆

Enfermedad de Rosai-Dorfman cutánea facial

Dear Editor:

Rosai-Dorfman disease (RDD) is a rare, benign histiocytic disorder characterized by generalized lymphadenopathy and constitutional symptoms, secondary to histiocytic infiltration of lymph nodes. Cutaneous Rosai-Dorfman disease (CRDD), the variant limited to the skin, is very rare. We present a case of CRDD on the left cheek that responded well to methotrexate.

A 38-year-old Moroccan man who had been living in Spain for about 6 years presented with a slow-growing asymptomatic lesion on the left cheek that had first appeared 18 months earlier and was not associated with any other symptoms. Physical examination revealed nonulcerated, yellowish, erythematous papules and nodules that formed a 4 cm infiltrated plaque (Fig. 1). The patient had no palpable lymph nodes or visceromegaly. Additional tests (complete blood count, biochemistry, erythrocyte sedimentation rate, C-reactive protein, angiotensin-converting enzyme, protein electrophoresis, immunoglobulins, complement, antinuclear antibodies, syphilis serology, hepatitis and human immunodeficiency virus (HIV), β2-microglobulin, and chest radiography) were normal or negative. Histopathologic examination revealed an unaffected epidermis and a granulomatous lymphohistiocytic inflammatory infiltrate (Fig. 2A) accompanied by plasma cells and few neutrophils in the dermis (Fig. 2B); histiocytes with abundant cytoplasm and vesicular nuclei showed striking phenomena of emperipolesis (intact inflammatory cells engulfed by histiocytes) (Fig. 3); and immunohistochemistry was positive for CD68 and S-100 protein and negative for CD1a. Microbiologic studies were negative for fungi, *Mycobacterium tuberculosis*, atypical mycobacteria and *Leishmania* spp. A diagnosis of CRDD was established on the basis of these findings and staging studies revealed no systemic involvement. Treatment was started with intralesional corticosteroids; partial response was achieved, so methotrexate (15 mg/wk) was added. Clear improvement was observed at 2 months.

RDD, or sinus histiocytosis with massive lymphadenopathy, is a form of non-Langerhans cell histiocytosis first described as a distinct entity in 1969 by Rosai and Dorfman[1–4]; it can occur in isolation or as part of other more complex conditions (R group histiocytoses).[5] RDD mainly affects young white and African American men and usually manifests as bilateral cervical lymphadenopathy associated with fever, weight loss, night sweats, fatigue,[5] leukocytosis with neutrophilia, and polyclonal hypergammaglobulinemia.[6] It is sometimes associated with autoimmune disorders such as lupus erythematosus, autoimmune hemolytic anemia, Crohn disease, primary cutaneous marginal zone lymphoma with IgG4 expression, and HIV infection. Extranodal involvement is present in 25% to 40% of cases. The skin, affected in up to 10% of cases, is one of the most frequently involved organs.[5,7] However, cases affecting only the skin are very rare.[2] Just over 100 exclusively cutaneous cases have been reported,[3,4,8] accounting for approximately 3% of all cases. Exclusively cutaneous cases most frequently affect middle-aged white and Asian women.

The clinical manifestations of CRDD are variable and nonspecific, including single or multiple papules, nodules, or plaques,[4] or, less frequently, other presentations such as pustules, acneiform lesions, and lesions mimicking vasculitis and panniculitis.[3] The face is the most common site, followed by the back, chest, thighs, hips, and shoulders. The presence of reddish-yellow nodules without tenderness to palpation can be useful in establishing a diagnosis.[3]

Histologically, the epidermis shows no abnormalities and a diffuse inflammatory infiltrate of histiocytes accompanied by lymphocytes, numerous plasma cells,[5] and isolated neutrophils is observed in the dermis. Phenomena of emperipolesis, an essential—although not pathognomonic—feature for diagnosis,[1] indicate that intact inflammatory cells and/or erythrocytes are present in intracytoplasmic vacuoles inside histiocytes, allowing them to be spared degradation by cytolytic enzymes,[1,3] in contrast to phagocytosis, in which the cells are destroyed. Nuclear atypia and mitotic figures are rare. Histiocytes are positive for S-100 protein and CD68 and negative for CD1a, which helps confirm the diagnosis and rule out other entities,[3] especially in extranodal lesions, which present a much lower frequency of emperipolesis.[1]

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